Non Syndromic Bilateral Dentigerous Cysts: An Unusual Case Report

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Abstract

Dentigerous cysts are odontogenic developmental cyst, which mostly surround the crown of unerupted teeth, odontomas, or supernumerary teeth. These cyst are mostly solitary in occurrence, their bilateral presentation is rare, especially in the absence of syndromes like Monteaux-Lamy, Basal cell nevus or Cleidocranial dysplasia. Non syndromic bilateral cysts show a predilection for the mandibular first molar area. We present a unusual case of a 11-year-old girl presenting with bilateral radiolucencies in associated with unerupted mandibular second premolars that were diagnosed histopathologically as dentigerous cysts .
KEYWORDS: Dentigerous cysts, Non Syndromic, odontogenic

Introduction

Dentigerous cyst or Follicular Cyst is Odontogenic, thought to be of developmental origin associated with the crown of an unerupted (or partially erupted) tooth. The cyst cavity is lined by epithelial cells derived from the reduced enamel epithelium and the enamel surface of the tooth forming organ (1). Regarding its pathogenesis, it has been suggested that the pressure exerted by an erupting tooth on the follicle may obstruct venous flow inducing accumulation of exudates between the reduced enamel epithelium and the tooth crown.

The Most Common Location of Dentigerous cysts are the Mandibular 3rd Molars and the Maxillary Canines . Dentigerous cysts are solitary,
Bilateral or multiple cysts are usually associated with developmental syndromes, such as mucopolysaccharidosis, basal cell nevus syndrome, and cleidocranial dysplasia.(2). The occurrence of bilateral dentigerous
cysts in the absence of a developmental syndrome is rare.(3),but their bilateral occurance in relation to mandibular premolars is very rare as in this case in absence of any syndrome.

In our knowledge only 24 cases of Non-syndromic bilateral dentigerous cysts are reported , out of which none of them were involved bilaterally in mandibular first premolar region.

Case report

A 11 year old girl reported to the Deparment of Pedodontics with the chief complaint of swelling on right and left side of the face (figure 1). She has noticed swelling about one and half months back. It was associated with intermittent pain. Extra oral examination revealed a diffuse swelling on the right side extending from 1 cm from corner of the mouth upto 3 to 4 cm anterior to posterior border of mandible. Superiorly it extended 1 cm below the ala tragus line and inferiorly upto the lower

Figure 1: Extra oral photograph showing asymmetry of the face bilaterally.

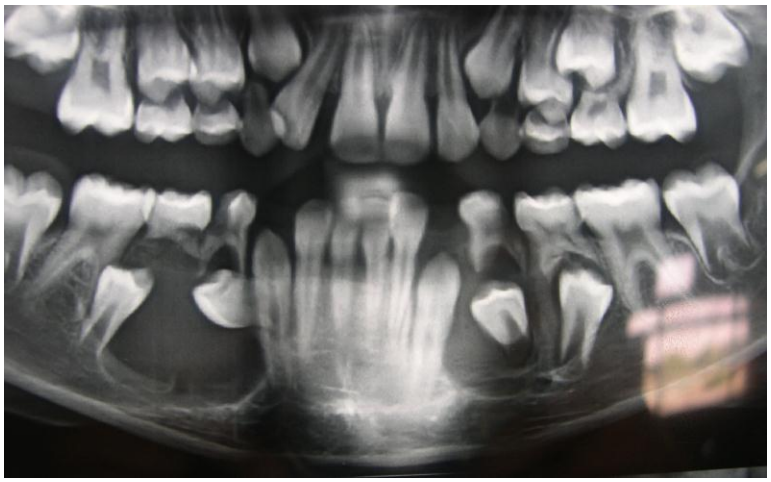

Figure 2: Panoramic radiograph showing two cyst like radiolucencies in relation to 44, 45, 34 & 35.

Figure 3: Intraoral Photograph showing swelling in relation to 44, 45, 34 & 35.

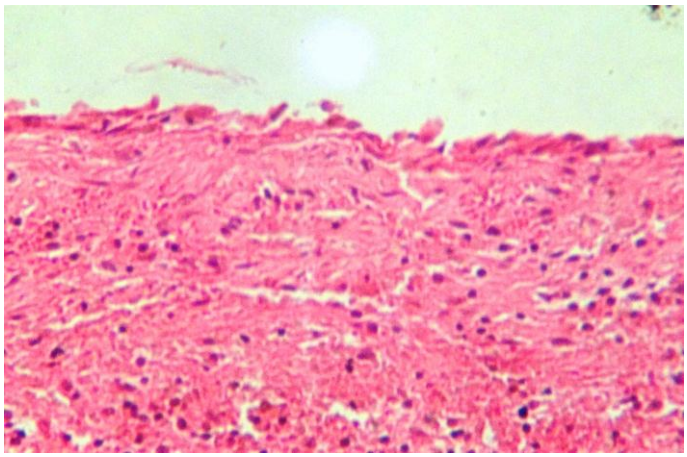

Figure 4: Photomicrograph of lesion showing features suggestive of dentigerous cyst (H&E, original magnification 40x) Right Side.

border of mandible(figure 2). Intraoral examination revealed a mixed dentition and clinically absent premolars and there was definite swelling in association with unerupted premolars i.e., 14 and 15 revealed hard bony swelling with obliteration of buccal and labial vestibule. Panoromic radiograph(figure 3) of the patient showed bilateral radiolucencies in the mandibular premolar region, on the right side a well defined unilocular radiolucency with sclerotic borders is seen beneath 84,85 and 33,extending lateral upto 43 till 46,44 and 45 are enclosed within the cystic cavity, pathologic root resorption of 84 and 85 were seen,radiolucency is also seen on left side which is well defined and unilocular enclosing 34,84 appears floating in the cavity.

Surgical removal of both the lesions was performed under general anesthesia along with removal of 84,85,74,44,and 34 , the specimens were submitted for histopathological examination. Histopathological examination revealed –tissues from both the cysts showed similar features. Cystic lumen was lined by a thin 2-3 cell layer thick epithelium with no rete- peg formation. The underlying connective tissue stroma was loose fibrocellular ; the fine collagen fiber bundles were dispersed with fibroblasts.The tissue was adequately vascular. Few chronic inflammatory cells were seen in addition to quiescent odontogenic islands. (figure 4,5)

Discussion

A dentigerous cyst is an epithelial-lined developmental cavity that encloses the crown of an unerupted tooth at the cementoenamel junction.(4). Dentigerous cyst occur more frequently in males (1.57:1) (5),.The present case was of an 11 year old female child .Dentigerous cyst are usually asymptomatic and occurs mostly in the 2nddecade and 3rd decade of life (6).It can become extremely large and cause cortical expansion and erosion, they can cause pain when they get infected. Radiographically, the dentigerous cyst appears as a unilocular radiolucency of variable size with well-defined

sclerotic borders, associated with the crown of an unerupted tooth. In an infected cyst the borders may be ill-defined. There may be difficulty in distinguishing a small cyst from a normal tooth follicle. It has been suggested that any follicular space of >5 mm should be suspected to be a dentigerous cyst(7).

In addition to the developmental origin, some authors have suggested that periapical inflammation of non-vital deciduous teeth in proximity to the follicles of unerupted permanent successors may be a factor for triggering this type of cyst formation as it was very evident in this case.

Histologically, a normal dental follicle is lined by enamel epithelium, whereas a dentigerous cyst is lined by non-keratinized stratified squamous epithelium. Since the dentigerous cyst develops from follicular epithelium it has more potential for growth, differentiation and degeneration than a radicular cyst. Occasionally the wall of a dentigerous cyst may give rise to a more ominous mucoepidermoid carcinoma. Due to the tendency for dentigerous cysts to expand rapidly, they may cause pathological fractures of jaw bones.

The usual radiographic appearance is that of a well-demarcated radiolucent lesion attached at an acute angle to the cervical area of an unerupted tooth. The border of the lesion may be radiopaque. The radiographic differentiation between a dentigerous cyst and a normal dental follicle is based merely on size. Radiographically, a dentigerous cyst should always be differentiated from a normal dental follicle. Dentigerous cysts are the most common cysts with this radiographic appearance.

Other lesions may mimic their radiographic appearance including, radicular cyst, and some odontogenic tumors like ameloblastoma, Pindborg's tumor, adenomatoid odontogenic tumor, calcifying odontogenic cyst, and ameloblastic fibroma.(1,7) In most instances, microscopic evaluation is therefore necessary to reach a definitive diagnosis. The treatment of a dentigerous cyst is usually dictated by the size of the lesion. If small and accessible it can be enucleated, but if the impacted tooth can erupt and if the lesion is large then marsupialization may be needed for the complete removal of a large cyst. Recurrence of a dentigerous cyst is rare and could be due to residual fragments of cyst lining. Although dentigerous cysts are commonly encountered, their bilateral occurrence has rarely been reported, especially in the absence of associated systemic disease or syndromes(8). Bilateral or multiple dentigerous cysts are usually encountered with Maroteaux-Lamy syndromeand Cleidocranial dysplasia or after prolonged concurrent use of cyclosporine and calcium channel blockers (9). Dentigerous cyst can attain considerable size with minimal or no symptoms, early detection and removal of the lesion is important to reduce morbidity.

Conclusion

Bilateral dentigerous cyst affecting mandibular first premolars are extremely rare and happens to be the first case reported in an non syndromic patient in english literature. An pedodontist who observes the patient in various stages of tooth development can contribute significantly by recognizing altered eruption patterns and can help in early diagnosis of pathology such as dentigerous cysts.

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